

# Serial casting for equinus deformity in children with cerebral palsy: A systematic review

Serial casting for equinus deformity

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## Abstract

**Aim:** The primary cause of gait deformity in children with cerebral palsy (CP) is equinus foot. Children with equinus foot typically receive physiotherapy as part of their care, as serial casting. In this study, we aimed to systematically examine the impact of serial casting procedure in children with CP with equinus deformity.

**Material and Methods:** Articles were identified from 2008 to 2019 by literature searches using Medline (MEDLINE), physiotherapy evidence base (PEDro), and Cochrane database. Randomized trials focused on CP children with equinus deformity were included. Data were extracted from the included studies and methodological quality of these data was evaluated using the PEDro scale. The modified Sackett scale was used to evaluate the level of evidence of each intervention.

**Results:** Four trials with good quality methodology were identified with good quality methodology. It was examined whether studies regarding intervention techniques were heterogeneous or not. Findings were analyzed in qualitative terms. This review revealed moderate evidence in three articles about the effectiveness of serial casting application and the ineffectiveness in one article, in addition to traditional physiotherapy programs. Meta-analysis was applied for homogeneous studies and it showed that serial casting could be used as a method to improve deformity of the equinus.

**Discussion:** The current systematic review analyzed four randomized controlled trials, applying strict inclusion selection criteria. The present evidence supports the use of serial casting for improving equinus deformity and modulates spasticity in children with CP and it is considered a moderate evidence of the efficacy of serial casting application. Although the findings of this review support the effectiveness of serial casting application in CP children with equinus deformity, additional randomized control trials with a larger sample size are still required to confirm the present evidence.

## Keywords

Serial casting; Deformity of equinus; Cerebral palsy; Toxin of botulinum; Abnormality of gait; Scissoring

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## Introduction

Equinus foot is a disorder of the musculoskeletal system often seen in patients with cerebral palsy (CP) and can restrict activities such as walking function [1]. Equinus is the most frequent problem in ambulatory children with spastic CP, resulting in an unstable and inefficient gait pattern. Without careful early treatment, it may evolve into permanent foot deformities which may require multiple surgical procedures. A variety of conservative approaches are currently available for managing Equinus in children with CP, such as bracing, stretching exercises, electrical stimulation, and casting/serial casting [2].

Through physical therapy, such as stretching and muscle strengthening exercises, antispastic medications, BTX-A injection, ankle-foot orthoses, and serial casting, or surgery, such as tendon elongation of Achilles, Equinus foot can be handled. BTX-A injection and serial casting are among such treatments with limited side effects relative to drug therapy [3]. Serial casting is described as using a series of progressive casts to increase the length of the muscle using low load prolonged stress on contracted tissues [4]. Serial casting aims to improve a pattern of the equinus gait in children with spastic CP (SCP) [5].

Numerous clinical trials have shown that casting is a useful adjunct to Botulinum toxin A (BoNT-A) for enhancing results, using either serial or single casts. In reducing spastic hypertonia, the combination of BoNT-A and casting was superior to BoNT-A alone [6]. This systematic review aimed at verifying the effect of the application of serial casting on equinus deformity for children with CP.

## Material and Methods

### Search strategy

This study was based on the recommendations of the Statement on Preferred Reporting Items for Systemic Reviews and Meta-Analysis (PRISMA) [7]. Eligibility criteria were defined as follows: (a) Participants: children with CP accompanied by equinus deformity aged from 2 to 17 years of both genders, (b) Interventions: the study group received serial casting after BoNT-A and traditional physiotherapy program, (c) Outcomes: Passive range of motion (PROM), spasticity degree, ankle kinematics during gait, gross motor function level and (d) Study design: Randomized controlled trials (RCTs). Anelectronic search was done from 2008 to 2019, in the Cochrane Central Register of Controlled Trials, PEDro and PubMed databases using the following keywords: “Serial casting” and/or “Equinus deformity” or “Cerebral palsy” or “Botulinum toxin” or “Scissoring” or “Gait abnormality”. The search was limited to RCTs only which published from 2008 to 2019. Studies were excluded according to the following criteria: (a) Non RCTs, (b) Participants aged over 17 years, (c) If no English translation is available and/or (d) Unpublished studies. Two authors independently evaluated each title and abstract identified in the search against the eligibility criteria. The full text was obtained for complete analysis.

### Data extraction

One reviewer extracted data from the included articles, and a second reviewer cross-checked it. The data extraction form included authors and year of publication, the characteristics of

the participant, measures of intervention, and outcomes [8].

### Quality assessment

Two authors applied the PEDro scale [9] separately to determine the quality of the trials and the third author resolved any disagreements.

### Data analysis

The following classification was used for the quantitative quality rating: PEDro score < 4 indicated poor quality; 4-5 indicated fair quality; 6-8 for good quality and 9-10 indicated excellent quality. The modified Sackett scale [10] was used for assessing the level of evidence as follows:

- Level 1a (Strong) = Well-designed meta-analysis or 2 or more ‘high’ quality RCTs (PEDro Scale scores  $\geq$  6) that show similar findings.
- Level 1b (Moderate) = One RCT of ‘high’ quality (PEDro Scale score  $\geq$  6).
- Level 2a (Limited) = At least one ‘fair’ quality RCT (PEDro Scale score = 4-5).
- Level 2b (Limited) = At least one well-designed non-experimental study: non-RCT; quasi-experimental studies; cohort studies with multiple baselines; single-subject series with multiple baselines.
- Level 3 (Consensus) = Agreement by an expert panel, a group of professionals in the field or a number of pre-post design studies with similar results.
- Level 4 (Conflicting) = Conflicting evidence of two or more equally designed studies.
- Level 5 (No evidence) = No well-designed studies: “Poor” quality RCTs with PEDro scores  $\leq$  3; only case studies/case descriptions or cohort studies/single subject series with no multiple baselines.

## Results

### Search results

The search identified 54 trials until July 2019. After screening titles and abstracts and removing duplicates, four studies (Dai and Demiryürek, 2017 [11]; Dursun et al., 2017 [12]; Kelly et al., 2019 [13] and Abd El-Monemet et al., 2019 [14]) were included in this review. Search results are presented according to the flow chart (Figure 1).

### Characteristics of the included studies

All included studies are RCTs. The summary of the included studies is presented in Table 1. The clinical homogeneity between some of the included trials allowed the quantitative analysis of their data.

### Qualitative analysis

#### Participants

The sample size ranged from 10 to 80. There were a total of 176 children participating across the four RCTs, diagnosed with CP, including both genders and aged from 2 to 17 years.

#### Interventions

The study versus the control groups in four of the included RCTs received botulinum toxin type A in addition to traditional sessions of physiotherapy.

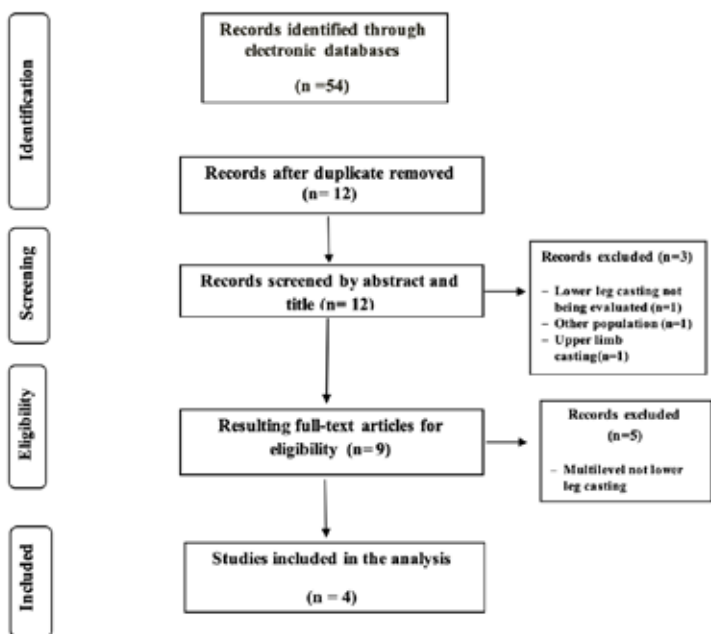
#### Outcome measures

In the reviewed studies, spasticity testing was performed using modified Ashworth scale (MAS) and modified Tardieu scale (MTS), passive range of motion (PROM) was assessed using

**Table 1.** Summary of the reviewed studies

	Dai and Demiryürek (2017) (11)	Dursunet al., (2017) (12)	Kelly et al., (2019) (13)	Abd El-Monem et al., (2019) (14)
Study design	RCT Level II	RCT Level II	RCT Level II	RCT Level II
Participants	Children with CP with scissoring of both legs	Spastic equinus foot due to CP	Children with spastic CP and equinus gait	Children with hemiplegic CP and equinus deformity
No. of participants	Control= 40 Study= 40	Control= 17 Study= 34	Control= 10 Study= 10	Control= 12 Study= 13
Intervention (study)	- BoNT-Ainjection and serial Casting in abductor position of the lower extremities. - Both groups received physiotherapy	- BoNT-Ato planter flexors and serial casting. - Both groups received physiotherapy for 3 weeks	- Injections into spastic Triceps surae muscles with Serial casting - Both groups received Physiotherapy	- 3 consecutive casts applied for 5 days each and removed in the last 2 days in each week to conduct the same physical therapy program
Intervention (Control)	BoNT-Ainjection without serial casting	BONT-A to Planter flexors without serial casting	Injections into spastic triceps surae muscles with single cast	Physical therapy program once a day/3times per week for 3 successive weeks
Outcome of interest	- Decrease spasticity - Reduce contracture due to spasticity - Gain additional ROM	- Decrease spasticity - Increased PROM - Improve gait	- Decrease of spasticity and hypo-extensibility of gastrocnemius-soleus complex - Improvement of PROM -Gross motor function improvement	- Reduction of spasticity Improvement of PROM - Improvement of ankle kinematic during gait
Measures	- GMFM 66 - MAS - CHQ	- MAS - MTS - OGS	- MTS - MAS - GMFM 66 -PEDI	- MAS - OGS
Component of health	Activity and participation	Activity and participation	Activity and participation	Activity and participation

RCT: Randomized control trial. BoNT-A: Botulinum toxin A. No: Number. ROM: Range of motion. PROM: Passive range of motion. GMFM: Gross Motor Function Measurement Scale. MAS: Modified Ashworth Scale. CHQ: Caregiver Health Questionnaire. MTS: Modified Tardieu Scale. OGS: Observational Gait Scale. PEDI: Pediatric Evaluation of Disability Inventory.



**Figure 1.** The PRISMA flow chart of the reviewed studies

**Table 2.** Methodology assessment of studies according to the Physiotherapy Evidence Database (PEDRO) scale

Criteria	Dai and Demiryürek (2017) (11)	Dursunet al., (2017) (12)	Kelly et al., (2019) (13)	Abd El-Monem et al., (2019) (14)
1-Specified eligibility criteria*	Yes	Yes	Yes	Yes
2-Random allocation of participant	Yes	Yes	Yes	Yes
3-Concealed allocation	No	No	No	No
4-Similar prognosis at baseline	Yes	Yes	Yes	Yes
5-Blinded participant	Yes	Yes	Yes	Yes
6-Blinded therapists	No	No	No	No
7-Blinded assessors	No	No	No	No
8-More than 85% follow-up for at least one key outcome	Yes	Yes	Yes	No
9-'Intention to treat' analysis	Yes	Yes	Yes	Yes
10-Between group statistical analysis for at least one key outcome	Yes	Yes	Yes	Yes
11-Point estimates of variability for at least one key outcome	Yes	Yes	Yes	Yes
Total PEDro score ( /10)	7	7	7	6
Quality	Good	Good	Good	Good

\*Item 1 does not contribute to the total score

(MTS), gross motor function level was assessed using GMFM-66 and gait abnormalities were assessed using observational gait scale (OGS).

**Quality of the included studies and the level of evidence**

The methodological quality of included four studies is presented in Table 2. The quality of the studies is good with a mean PEDro score (range 6 to 7). The four included studies had similar groups at baseline, analyzed the between-group difference. The four studies carried out an intention-to-treat analysis, none of the studies blinded participants.

**Evidence of serial casting application interventions**

The results of the 4 reviewed trials (Dai and Demiryürek,2017, Dursun et al., 2017, Kelly et al., 2019, and Abd El-Monemet al., 2019) [11-14] which investigated the effects of serial casting application for CP children with equinus deformity are summarized in Table 3.

**Statistical analysis:**

Only three articles (Dai and Demiryürek (2017), Dursun et al., (2017), Kelly et al., (2019)) [11-13] have homogeneity in all four components (participants, intervention, outcome (modulation of spasticity) and the outcome measures) so only one meta-analysis was performed and this meta-analysis favors the use of serial casting application for modulation of spasticity in children with spastic CP.

**Description and Interpretation of Forest plot of RevMan:**

The Forest plot is composed of (from left to right):

1. The names of the included studies.
2. The data of the study and control groups including the mean difference between pre- and post-intervention, SD of the paired

**Table 3.** Summaries means of study groups and control groups and the difference between this means

	Dai and Demiryurek (2017) (11)	Dursunet al., (2017) (12)	Kelly et al., (2019) (13)	Abd El-Monem et al., (2019) (14)
Outcomes	- Decrease spasticity - Reduce contracture due to spasticity - Gain additional ROM	- Decrease spasticity - Increased PROM - Improved gait	- Decrease of spasticity and hypo-extensibility of gastrocnemius-soleus complex - Improvement of PROM - Gross motor function improvement	- Reduction of spasticity - Improvement of PROM - Improvement of ankle kinematic during gait
Means of the control group	- GMFM-66 mean: Pre= 42.26 Post= 64.57 - MAS mean: Pre= 3.81 Post= 2.78 - CHQ mean: Pre= 44.29 Post= 64.63	- MAS mean: Pre= 4 Post= 3.1 - MTS mean: Pre= 79.4 Post= 83.5 - OGS mean: Pre= 8.5 Post= 9.5	- MTS mean: Pre= -3.6 Post= 12 - MAS mean: Pre= 2.6 Post= 1.3 - GMFM-66 mean: Pre= 72.7 Post= 76.5 - PEDI mean: Pre= 74.6 Post= 81.2	
Means of the study group	- GMFM-66 mean: Pre= 41.18 Post= 77.37 - MAS mean: Pre= 3.74 Post= 1.88 - CHQ mean: Pre= 42.66 Post= 75.95	- MAS mean: Pre= 4 Post= 2.4 - MTS mean: Pre= 79.8 Post= 93 - OGS mean: Pre= 7.5 Post= 10.5	- MTS mean: Pre= -3.7 Post= 9.4 - MAS mean: Pre= 2.8 Post= 0.9 - GMFM-66 mean: Pre= 77.4 Post= 80.5 - PEDI mean: Pre= 77.3 Post= 82.1	
Differences between means	- GMFM 66: Control group=22.31 Study group= 36.19 - MAS: Control group=-1.03 Study group= -1.86 - CHQ: Control group=20.34 Study group= 33.29	- MAS: Control group=-0.9 Study group=-1.6 - MTS: Control group= 4.1 Study group= 13.2 - OGS: Control group= 1 Study group= 3	- MTS: Control group= 15.6 Study group= 13.1 - MAS: Control group=-1.3 Study group=-1.9 - GMFM 66: Control group= 3.8 Study group= 3.1 - PEDI: Control group= 6.6 Study group= 4.8	- OGS: Control group= 7.16 Study group= 10.07

PROM: Passive range of motion. GMFM: Gross Motor Function Measurement Scale. MAS: Modified Ashworth Scale. CHQ: Caregiver Health Questionnaire. MTS: Modified Tardieu Scale. OGS: Observational Gait Scale. PEDI: Pediatric Evaluation of Disability Inventory.

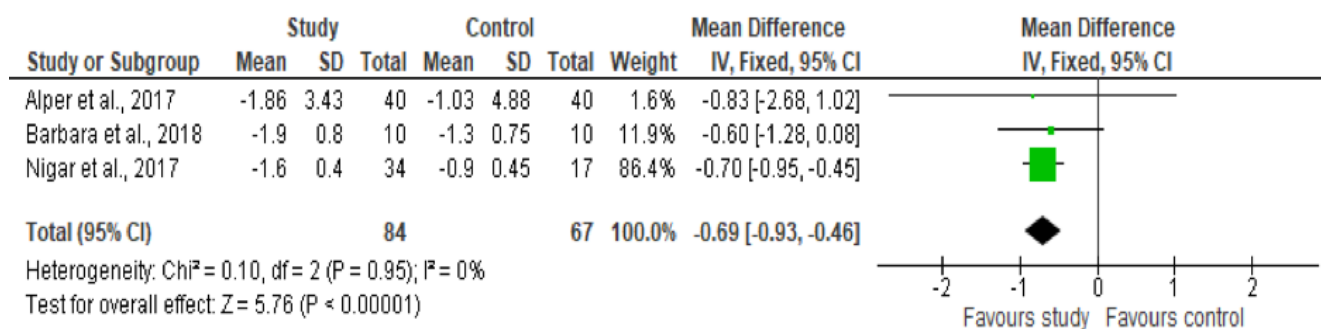
difference, and number of cases in each group.

- The weight of each study as a % of the total of the meta-analysis (100%).
- The difference in means between the 2 groups + the 95%CI of the difference.
- The publication year again.
- On the right side, there is a figure showing the same above results. Each study is represented by a square (its size = study weight, and its center is opposite to the mean's difference) on a straight line (representing the 95%CI of the mean's difference). The final results of the meta-analysis are represented by the black diamond (its center is the mean's difference across all studies and the tips are the 95%CI of the mean's difference across all studies).
- The line in the middle of the graph is opposite the 0 value and it is called the equator line which means no difference between the groups. If the lines of any study and/or the diamond touch it, this means that there is no statistical difference between the 2 groups.
- The last 2 lines written in the plot are for the heterogeneity represented by I2 statistic as a % and a p-value. When the p-value is < 0.05, then heterogeneity is considerable across the studies and we should take the results cautiously. The 2nd line is the p-value of the overall results (that are represented by the diamond in the graph). This is represented by Z- value and p-value. When the p-value is < 0.05, this means that the overall result is statistically significant.

9. Forest plots were done using Review Manager (RevMan) [Computer program] Version 5.3. Copenhagen: The Nordic Cochrane Centre, the Cochrane Collaboration, 2014.

**Statistical methods:**

We analyzed data from the included studies using Review Manager (RevMan – version 5.2, The Nordic Cochrane Center, The Cochrane Collaboration, Copenhagen, Denmark), and Microsoft Excel 2010 (Microsoft Corp., Redmond, WA, USA). A formal meta-analysis was conducted for our outcome (difference in MAS between pre- and post-intervention). The pooled MAS values were expressed as the mean difference (MD) between study and control groups, with 95%CI. We explored and quantified between-study statistical heterogeneity using the I2 test. Since I2 was 0%, we used the fixed-effect model. We considered 2-sided statistical analysis testing setting the  $\alpha$ -error level at 0.05 (Figure 2).



**Figure 2.** Forest plot of comparison: 1 Comparison between study and control groups, outcome: 1.1 Difference in modified Ashworth scale between pre and post interventions.

## Discussion

The aim of the current review is to determine the efficacy of serial casting on the deformity of the equinus in children with CP. The analysis covers studies conducted between 2008 and 2019 searched via PubMed, PEDro, Cochrane library, and Google scholar databases in the Medline database that most likely contain a large number of papers published annually.

The current systematic review analyzed four randomized controlled trials, applying strict inclusion selection criteria. All trials satisfied at least six PEDro-scale criteria. Three studies, including Dai and Demiryürek (2017) [11], Dursun et al. (2017) [12] and Kelly et al. (2019) [13] underwent meta-analysis.

The scoring of each study of the three studies with the PEDro scale is seven after collecting data based on AACPD sheet items. The higher the number of scores of factors measuring the study's efficiency, the greater the study's efficiency. The research design of the three studies is randomized controlled trials with evidence level two; children involved in the three studies were spastic CP with ages ranging from 2 to 17 years. The age range in Dai et al.'s study (2017) [11] was 2-4 years, in Dursun et al.'s study, (2017) [12] was 3-17 years and in Kelly et al.'s study (2019) [13], was from 2-7 years.

In the study by Dai and Demiryürek (2017) [11], the intervention was a 3-week serial casting application once a week and consisted of a dual cast which consisted of a standard long leg plaster cast and a circular cast from below the knee to above the knee, while in the study by Dursun et al. (2017) [12], the intervention was three consecutive serial casting applications for three weeks once a week and consisted of a dual cast which consisted of a standard short leg plaster cast and a circular cast from below the knee to above the knee, with a follow-up period of 12 weeks. Moreover, the intervention in the study by Kelly et al. (2019) [13] was six weeks once a week and consisted of a dual cast which consisted of a standard short leg plaster cast.

Dai and Demiryürek (2017) [11] compared the use of serial casting with botulinum toxin type A in children with CP and spastic Paraparesis with scissoring of lower limbs. Dursun et al. (2017) [12] compared the effectiveness of intermittent serial casting on equinus foot in children with CP and BoNT-A. Kelly et al. (2019) [13] compared the effect of serial casting in spastic hypertonia of the triceps surae muscle with CP and equinus foot and BoNT-A.

All three studies used MAS to assess spasticity. Dai and Demiryürek (2017) [11] used GMFM to assess the motor function of their participants. Dursun et al. (2017) [12] used MTS to assess spasticity and OGS to assess improvement of gait. Kelly et al. (2019) [13] used MTS to assess spasticity, GMFM to assess the motor function of their participants, and PEDI for assessing social function. The present evidence supports the use of serial casting applications for improving equinus deformity in children with CP.

In the study by Dai and Demiryürek (2017) [11], GMFM and child health questionnaire scores (CHQS) were markedly elevated in both groups following Botulinum toxin type A treatment, MAS scores decreased for 12 weeks in the Botulinum toxin type A injection with serial casting group ( $p < 0.05$ ), however no marked reduction in MAS scores in the Botulinum toxin type A injection

only group.

In the study by Dursun et al. (2017) [12], there was a significant improvement in MAS, PROM in both groups ( $p < 0.001$  for all parameters of each group). The mean MAS and PROM of the casting group were better than those of the control group at week 4 ( $p = 0.006$ ,  $p = 0.013$ ) and week 12 ( $p = 0.015$ ,  $p = 0.013$ ) with significant improvement in OGS in both groups ( $p < 0.001$  in both groups).

Regarding Kelly et al.'s study (2019) [13], there is a significant improvement of MTS of dorsiflexion with knee extension ( $p < 0.001$ ) and dorsiflexion with knee flexion ( $p < 0.001$ ), a significant improvement for GMFM-66 ( $p = 0.002$ ) and all PEDI domains except social function caregiver assistance ( $p = 0.009 - < 0.001$ ).

There was no significant difference in ankle ROM, the distribution of spasticity grades of calf muscle tone, and the total score of OGS ( $p = 0.64$ ,  $p = 0.44$ ,  $p = 0.64$ ) in Abd El-Monem et al. (2019) [14].

### Conclusion:

Although results from this analysis support the efficacy of using serial casting application for children with CP associated with equinus deformity, there is still a need for additional RCTs with larger sample sizes to validate the evidence.

### Scientific Responsibility Statement

*The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.*

### Animal and human rights statement

*All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.*

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### Conflict of interest

*None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.*

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